Beauty parlor stroke syndrome due to a bone fragment from an osteophyte of the atlas: case report

Takuya Kameda, MD,1 Koji Otani, MD, DMSc,1 Takamitsu Tamura, MD, PhD,2,3 and Shinichi Konno, MD, PhD1

Departments of 1Orthopaedic Surgery and 2Neurosurgery, Fukushima Medical University, Fukushima, Japan; and 3New England Center for Stroke Research, University of Massachusetts Medical School, Worcester, Massachusetts

Beauty parlor stroke syndrome (BPSS) is a rare condition characterized by mechanical impingement of a vertebral artery (VA) during neck rotation and/or hyperextension followed by vertebrobasilar insufficiency. However, there have been no reports of BPSS in which the cause of mechanical impingement was identified and no cases for which surgical treatment was reported. The authors report the case of a 56-year-old Japanese man who presented with presyncope that occurred during cervical extension. Given the possibility of vertebrobasilar insufficiency, digital subtraction angiography and CT angiography were performed. These studies revealed that the right VA was hypoplastic and the left VA was dominant. Moreover, in the position of cervical extension, the dominant left VA showed constriction caused by a bone fragment of an osteophyte of the atlas. Removal of the bone fragment was performed. Postoperative left vertebral angiography showed improvement of blood flow in the extended position, and the presyncope completely disappeared. The pathomechanism of this case was a bone fragment compressing the left VA in the C-1 groove during neck extension. In BPSS patients with recurrent transient symptoms, the possibility of this mechanism of VA constriction by a free bone fragment should be considered.

https://thejns.org/doi/abs/10.3171/2017.7.SPINE17226

KEY WORDS beauty parlor stroke syndrome; cervical extension; vertebral artery; bone fragment; bow hunter’s syndrome; adjacent-segment degeneration

Beauty parlor stroke syndrome (BPSS),18 which is caused by the development of vertebrobasilar ischemia and infarction,6,21 has recently become well known.7 Various symptoms—sensory disturbance, weakness, dizziness, tinnitus, headache, nausea, hemiparesis, and nystagmus16—have been reported in individuals who had their head hanging backward while seated and having their hair shampooed at a beauty salon. Thus, the pathogenesis of BPSS has been suggested to be mechanical impingement through neck rotation and/or hyperextension, resulting in vertebral artery (VA) blood flow reduction at the atlanto-occipital (O–C1) junction.18

Although there have been several case reports of BPSS, there are no published papers explaining the underlying mechanism or describing surgical treatment. Here we report a case of BPSS in which the pathological mechanism of BPSS could be determined. Surgical removal of the apparent causal factor relieved the patient of his symptoms and prevented further infarction.

Case Report

History and Presentation

A 56-year-old Japanese man presented with the chief
complaints of episodes of presyncope, unconsciousness, and a wobbly feeling. He had a history of measles at 3 years of age, after which he developed a symptomatic state similar to athetoid-type cerebral palsy, with involuntary motion and hypertonia. However, he had managed to remain independent, living on his own. Seven years prior to onset of the current symptoms, he had undergone cervical spinal fusion (C2–T2 posterior, C5–6 anterior) for cervical myelopathic symptoms caused by his involuntary motions. Four years prior to the present admission, the C-2 screws were removed because they were causing neck pain. From 1 year prior to this admission, when the patient extended his cervical spine, he felt faint, became wobbly in his gait, and feared that he would “black out.”

On admission, the patient’s muscle strength was assessed with manual muscle testing and given a grade of 4/5 for the extension strength of the left wrist and left fingers. Testing of his deep tendon reflexes revealed a reduction of the left biceps reflex and left brachioradial reflex, with an exaggeration of the lower limb deep tendon reflexes. No sensory disturbance was found. These neurological findings had not changed from the time of the patient’s last operation. He reported that when extension of the cervical spine caused him to feel faint, returning to a neutral position relieved this symptom immediately, and that the wobbly sensation appeared only when he stood up in conjunction with cervical extension. We were able to reproduce these phenomena on examination in our outpatient clinic. Based on these findings, the possibility of vertebralbasilar insufficiency was considered.

Imaging Studies

Radiographs of the cervical spine showed atlantoaxial translation in the flexed position: the atlantoaxial interval was 3.9 mm (Fig. 1). During maximum extension, the angle between the occipital bone and the atlas was 40.3° based on the method by Yoshimoto et al. This angle was much larger than the normal angle of extension, which is approximately 15°. The position of the occipital bone was slightly shifted posteriorly during cervical extension. MRI of the brain revealed a high-intensity area in the right cerebellum on T2-weighted images (Fig. 2). There was no high-intensity area on diffusion-weighted images. A previous infarction of the cerebellum was suspected.

Computed tomography angiography (CTA) showed no VA interruption in flexion (Fig. 3A and B). In extension, however, interruption of the left VA was detected. This interruption was caused by compression between the inferior rim of the C-1 groove and a bone fragment from an osteophyte, i.e., the posterior osteophyte of the fovea articularis superior atlantis (Fig. 3C and D). Digital subtraction angiography (DSA) was performed with injection of both internal carotid arteries (ICAs) and both VAs (4-vascular DSA) and revealed hypoplasia of the bilateral posterior communicating arteries (PCoAs) (Fig. 4A and B) and no filling of intracranial vessels upon right VA injection (Fig. 4C). The intracranial posterior circulation was thus being supplied exclusively from the left VA (Fig. 4D). Dynamic left VA angiography in the extended position showed constriction of the VA at the atlanto-occipital junction (Fig. 5B), although the blood flow of the artery was sufficient in the neutral position (Fig. 5A).

Based on the above-described findings, we concluded that the left VA (the dominant VA) was constricted by the bone fragment and that the reduced blood flow was the cause of the patient’s bouts of unconsciousness. A decision was made to remove the bone fragment surgically.

Operation and Postoperative Course

The operation was performed under general anesthesia. To maintain the cervical flexion position during the operation, the patient’s head was fixed in the prone position with a head frame. The free bone fragment was removed (Fig. 6), and an inferior rim osteotomy of the C-1 VA groove was also performed to remove the portion of the atlas that was contributing to the bony compression. Because of the paravertebral venous plexus around the V3 portion of the extracranial VA, we were not particularly concerned about mechanical stress on the VA, which can lead to stroke and vasospasm, when removing the pathological free bone and inferior rim of the C-1 groove. Therefore, no medication was administered for stroke or vasospasm prophylaxis, and these conditions did not occur. Postoperatively, disappearance of vessel compression in the extended position was confirmed by angiography (Fig. 5D). The patient’s chief complaint of presyncope disappeared completely.
During 4 years of follow-up, he has had no recurrences. In radiographs obtained at the final follow-up visit, the angle between the occipital bone and atlas at full extension was similar to that before surgery.

Discussion

In this report we have described a case of BPSS caused by a bone fragment from an osteophyte compressing the VA. Removal of the bone fragment relieved the patient’s presyncope as well as radiographic evidence of VA compression, and the patient has had no further infarction.

Bone fragment should thus be considered as one of the potential causes of VA constriction in other BPSS patients. Some hypotheses about the details of mechanical impingement have been proposed—namely, the impingement has been attributed to a downward shift of the contralateral atlas with a fixed VA segment or compression between the occiput and the vertebral arch of the atlas during prolonged hyperextension. Nonetheless, to our knowledge, there has been no report directly demonstrating impingement as a causative factor in a BPSS case. In the present case, the anatomical location revealed by CTA demonstrated that the free bone fragment came from the posterior osteophyte of the fovea articularis superior atlantis. We speculate that when the patient’s neck was extended, the posterior bone fragment was pushed by an occipital condyle and became stuck in the groove for the VA, and the vessel was compressed as a result.

This case also demonstrated that that chief compliant of BPSS could be resolved and that an expected infarction could be avoided by removing the constricting bone fragment. In this patient, the impingement factor was identified by CTA with cervical extension, and removal of the bone fragment was performed according to the CTA findings. Fusion of the occipital bone and the axis was not performed in the present case because we decided that the patient’s range of cervical motion should be retained as much as possible. To our knowledge, there have been no reports on the surgical treatment of BPSS. Moreover, a previous infarction was confirmed by MRI in the present case, but no further infarction developed. Therefore, in patients in whom an apparent impingement factor is detected, surgical release could be an effective treatment to relieve symptoms and prevent further infarction.

FIG. 3. CTA of the cervical region. In flexion (A, coronal, and B, left sagittal view), the left VA could be observed through the groove of the atlas. In extension (C, coronal, and D, left sagittal view), the flow of contrast medium in the left VA is interrupted by a bone fragment (arrows). Based on the results shown in C and D, the angle of the O–C1 joint is wider in extension than in flexion.
Our patient’s chief complaint was presyncope, which is not among the typical BPSS symptoms (i.e., sensory disturbance, weakness, dizziness, tinnitus, headache, nausea, hemiparesis, and nystagmus). DSA in the present case revealed that the left VA supplied both sides of the posterior intracranial circulation and that there was no collateral blood flow from the bilateral PCAs. We thus concluded that once the dominant left VA was occluded, broad ischemia developed, followed by presyncope. This temporary symptom was similar to bow hunter’s syndrome (BHS) except for the triggering position.

BHS is caused by vertebrobasilar insufficiency induced by head rotation. It is mainly the result of underlying compression due to osteophytes and/or segmental instability and characterized by transient symptoms, such as a presyncopal sensation, syncope, vertigo, dizziness, nausea, and/or emesis. Thus, BPSS and BHS could have consecutive, collinear pathogenetic mechanisms. From this viewpoint, the management guideline for BHS—in which the gold standard of diagnosis is dynamic angiography, with decompression being the appropriate treatment when VA compression is observed—could be useful in some BPSS patients who experience recurrent symptoms and in whom the compressing factor can be detected. However, since BPSS also includes pediatric cases and trauma cases, caution should be exercised when selecting the treatment.

Our patient had undergone cervical spinal fusion (C2–T2 posterior, C5–6 anterior) prior to experiencing BPSS. Dynamic radiographs showed an extreme range of motion of the atlanto-occipital (O–C1) joint (reaching 41°), which could indicate posterior sliding of an occipital condyle. Thus, there is a strong possibility that the bone fragment following the fracture of the osteophyte was the result of stress on the O–C1 and C1–2 joints caused by the fusion of C2–T2. Additionally, this patient had a symptomatic state with involuntary motion and hypertonia. In other words, while the specific intervertebral level immediately superior to the fused levels was C1–2, one of the pathological mechanisms of BPSS may be the same as in adjacent-segment degeneration (ASD) given that the only mobile joints for cervical flexion and extension were O–C1 and C1–2.

Biomechanical investigation has demonstrated that fusion increases the range of motion of the adjacent level, potentially accelerating degenerative changes and leading to ASD. Our patient’s pathology may be understood as a complication of the cervical fusion. With a long fusion, as in the present case, it should be considered that the BPSS could be caused by biomechanical stress on the joints that are still mobile.

As shown by this case, the existence of a bone fragment from an osteophyte can cause BPSS. In such cases, removal of the bone fragment is required. BPSS patients with temporary symptoms should be evaluated carefully, and the most suitable treatment for the relief of symptoms and prevention of further symptoms should be identified. No guideline regarding the diagnosis and therapeutic strategy for BPSS has been established, and an accumulation of further cases is necessary.
T. Kameda et al.

References

15. Piñol I, Ramirez M, Saló G, Ros AM, Blanch AL: Symptomatic vertebral artery stenosis secondary to cervical...

Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: all authors. Acquisition of data: all authors. Analysis and interpretation of data: all authors. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Konno. Administrative/technical/material support: all authors. Study supervision: Otani.

Correspondence
Koji Otani: Fukushima Medical University, Fukushima, Japan. kotani@fmu.ac.jp.